ISSN: 2577-2996

PROGRESS IN MEDICAL SCIENCES, 2023
VOL 7, NO. 2, PAGE 1 – 4
DOI: doi.org/10.47363/PMS/2023(7)187

CASE REPORT

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Acquired Intrabdominal Testis Due to Adhesions After Necrotizing Enterocolitis

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ABSTRACT

Un descendent testis (UDT) refers to absence of the testicle in the hemiscrotum. It happens usually due to the stagnation of the testicle during the descent. In about 90% of cases is unilateral. The etiopathogenesis of UDT is quite well described in the literature where in most cases stagnation occurs in the inguinal canal and very rarely in the abdomen. Stagnation of a testicle in the abdomen in addition to the histopathological damage that may result, may also be the target of adhesions with intestinal segments. There have been documented some very rare reports of intestinal obstruction due to adhesions between an intraabdominal testis and the intestinal segment. Our case report aims to document a very rare variant of acquired intra-abdominal cryptorchidism due to adhesions after necrotizing enterocolitis (NEC). Any newborn after a laparotomy has a significant risk of developing intraperitoneal adhesions, which in the NEC are common complications even when passed without surgical treatment. In this report we will describe a case of previously palpable testis in the inguinal canal at neonatal age which was pulled inside the abdomen at seventh month of age through adhesions between test and the segment of sigmoid colon after NEC.

ARTICLE HISTORY

Received February 15, 2023
Accepted February 18, 2023
Published April 01, 2023

Keywords: Un Descendent Testis, Acquired Cryptorchidism, Necrotizing Enterocolitis, Adhesions, Newborn, Laparotomy

Introduction

Undescended testis (UDT) refers to the absence of the testicle in the hemiscrotum mainly due to stagnation of the testis during descent. It is among the common causes of surgical interventions in pediatric surgery. It is estimated that 1-2% of male babies have UDT till the 12th months of age. In most cases stagnation occurs in the inguinal canal and very rarely in the abdomen where, in addition to the histopathological damage that may result, it may also be the target of adhesions with intestinal segments. There have been documented some very rare reports of intestinal obstruction due to adhesions between an intraabdominal testis and the intestinal segment. In the newborn age, the most common cause of adhesions is necrotizing enterocolitis (NEC) with consequently ileostomies. Although obstructive ileus as a consequence of this adhesions between bowel segments is well known, intestinal obstruction due to adhesions between testicular structures and bowel segments has been reported very rarely. Our case documents a very rare cause as an etiological factor of acquired intraabdominal cryptorchidism due to adhesions after NEC. Recurrent cryptorchidism refers to a testis that was previously much more in the lower line to descend in scrotum but had reascend-retract in the upper line due to different factors. While the pathogenesis of congenital undescended testis is considered multifactorial including hormonal, genetic, and environmental influences the etiology of recurrent cryptorchism remains unclear [1-4]. Acquired cryptorchidism has a reported prevalence of 1%–7% and peaks around 8 years of age [5,6]. We have reviewed the literature and to our knowledge this is first case reported where adhesions due to NEC have been cause of acquired intrabdominal testis in infant age.

Case

we will describe a case of a testis previously palpable in the inguinal canal at neonatal age that was retracted intraabdominally after seven months of age due to adhesions between the testis and the sigmoid colon segment after NEC. He was operated on at neonatal age from perforated bowel segment due to complicated NEC. An ileostomy was performed, which was closed three months later. At the time of the first operation when the ileostomy was performed as well as three months later when the ileostomy was closed, the presence of the testis was noted in the inguinal canal. The patient at the time of the first operation had many adhesions that were released, which were much more evident

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when the ileostomy was closed. While under anesthesia, the patient was examined for the undescended testis, and it was found to be in the inguinal canal and could reach the neck of the hemiscrotum with slight retraction. After closure of the ileostomy, the patient was discharged on the 5th postoperative day and was followed in our outpatient clinic every 2 months, including for the left undescended testis with ultrasound and physical examination. At the first outpatient visit at five months of age, the left hemiscrotum showed the absence of testicle which was slightly smaller than the right testicle and was evident in the inguinal canal while the contralateral testicle was normal. At the second visit, two months later, it was evidenced that the left testis has retrieved into the internal inguinal orifice and it was not able to pull it near the neck of the left hemiscrotum, as it was possible in the previous visit. At the third visit when the child was nine months old, we could not palpate the testis in the inguinal canal. On ultrasound it was found to be close to the internal inguinal orifice and the same was confirmed

by pelvic MRI and was smaller than contralateral (Figure 1).

Figure 1: MRI of the pelvic demonstrated the presence of left testis in the pelvis, near the left inguinal canal.

The child was scheduled two months later for laparoscopic exploration and was found to have testis attached by adhesions to the loop of the sigmoid colon (Figure 2).

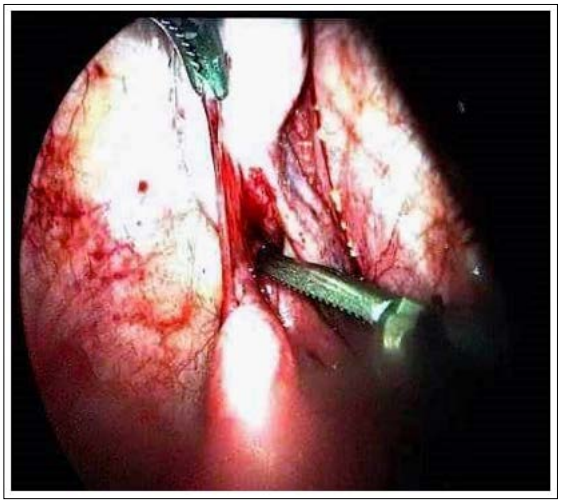

Figure 2: Intraoperativ picture during laparoscopy demonstrates adhesions of sigmoid loop with the left testis.

Adhesions and peritoneal attachments were released to gain length and the testis was brought to the neck of the hemiscrotum with slight tension where it was attached. Patient was discharged the following day. Five months after laparoscopic orchiopexy, the testis remains in the neck of the left hemiscrotum with good vascularization but with very small progressive development (10x6mm) compared to the contralateral testis (11.5x7.5 mm) (Figure 3).

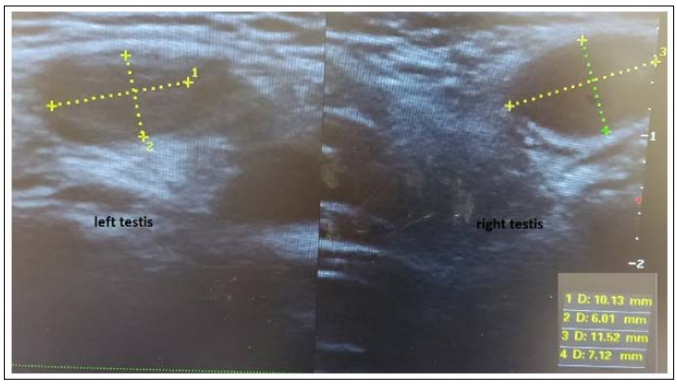

Figure 3: Ultrasonographic image of the left and the right testis, 5 months after laparoscopic orchiopexy

Discussion

Bowel obstruction due to adhesions are seen in children usually after laparotomies. NEC is often cause of adhesions which can occur even when passed without need for laparotomy. These adhesions usually occur between bowel segments and with surrounding intraperitoneal structures also. UDT can be target of these adhesions when still intraabdominally but adhesions have not been prescribed to be cause of acquired intra-abdominal testis as occurred in our case. On the other hand, cryptorchidism is defined as a failure of a testis to reach scrotum. It occurs in 3% of full-term and 30% of preterm male neonates. However, above 80% of this cases descent spontaneously by the 12th month of age [7-8]. After this age has to be operated due to histological changes that start to occur. UDT is associated with increased risk for torsion, trauma, infertility, malignancy and has the psychological impact of an absent testis later on adulthood also [1,2]. The risk of tumors (seminoma) in cryptorchid testes is 10 to 40 times higher than in descendent one [10,11]. Also, acquired cryptorchidism as well as congenital UDT are at risk for developing the same adverse histologic changes seen in primary cryptorchid testes requiring surgical correction [12]. Our case had a mild hypotrophy probably due to hypoperfusion provoked by vascular stretching through the sigmoid segment. About 10% of testicular torsion occurs in UDT, also bowel obstruction has been documented to occur from adhesions between the intra-abdominal testis and bowel [13,14]. Our case clearly shows the correlation between NEC and acquired intra-abdominal testis where adhesions due to NEC attached testis to the sigmoid colon segment and retracted the testis inside the abdominal cavity. In our case the adhesions were responsible for testicular retraction from the inguinal canal into the abdominal cavity. About 20% of cases of undescended testicles cannot be palpated on regular examination. A quarter of these cases can be palpated when the child is under anesthesia, and 6% of non-palpable cases are intraabdominal [7]. Also, it is well known that retractile testes are at risk of becoming true ascended testes, so yearly examination is recommended [15,16]. The case we have described here was positioned in the inguinal

canal in the first five months, then ascended into the abdomen due to retraction from adhesions attached to the sigmoid colon ending in the intraabdominal testis. Our case clearly shows the correlation between NEC and acquired intra-abdominal testis where adhesions due to NEC attached the testis to the segment of the sigmoid colon and retract testis from the inguinal canal into the abdominal cavity. This case highlights the importance of measures that need to be taken during adhesiolysis when a testis is in inguinal canal and can easily slide back inside the abdominal cavity and potentially becoming adherent in bowel segment due to adhesions thus potentially causing acquired intraabdominal testis. Cryptorchid testes undergo parenchymal changes if surgical correction is delayed beyond 2 years of age [17]. Our case showed slow development and was noticed to be slightly hypotrophic. Imaging exploration of the pelvic cavity in case of unpalpable testis is mandatory especially when one was previously evidenced to be in inguinal canal since it can become torqued and atrophied or even retract intraabdominally like it happened in our case. If during surgical intervention, testis is found to be severely hypoplastic, gonadal excision has to be considered [17]. Preoperative diagnosis allowed us to have a discussion with parents for what they decided to perform orchiopexy even knowing from imaging studies that testis was slightly hypotrophic. Any neonate requiring a laparotomy have a significant risk for developing adhesions [18]. In case of inguinal testis, it is important to explore the region near the inguinal canal for assuring not to leave any potentially adhesions possibility that can attach testis and latter retract it in abdominal cavity like it has happened in our case after NEC. Our case emphasizes a very rare presentation of acquired intra-abdominal testis. Further, delineates the importance of thorough intraoperative exploration of internal inguinal ring during laparotomies especially when pathologies like NEC are followed with adhesions and when a testis is in inguinal canal or even absent. However, few cases have been reported showing correlation between cryptorchidism and intestinal obstruction in children [18,19]. Reviewing the literature, to our knowledge acquired intrabdominal testis in children caused after NEC adhesions has not been reported previously.

Conclusion

As our case emphasizes a very rare etiologic factor of acquired intra-abdominal testis, we think that it need to be estimated and should be reported in literature in context of etiopathology of UDT eventually.

Pediatric surgeons should consider as risk factor for acquired intra-abdominal testis when a neonate or infant with inguinal testis undergoes laparotomy especially after diseases with potentially adhesions production such as NEC.

Thus, is mandatory if an infant with UDT in inguinal canal undergoing laparotomy especially from the diseases with high risk for adhesions to take measures for preventing adhesions near inguinal canal as a potentially risk factor for acquired intra-abdominal testis.

Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

Conflict of Interest

The following authors have no financial disclosures: (IsberAdemaj, Hysni Jashari, Naser Gjonbalaj and Arta Syla).

Authors Contributions

Isber Ademaj carried out design of the study, data collection, drafted the manuscript and was a major contributor in writing the manuscript supervision and analysis of the literature. Hysni Jashari and Arta Syla performed the literature review and was involved in writing the manuscript. Naser Gjonbalaj performed radiological examination of the pelvic MRI. All authors read and approved the final manuscript.

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